Case Reports

Mycotic Aneurysm and Disseminated *Mycobacterium avium-intracellulare* Infection in a Patient With Hairy Cell Leukemia

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HAIRY CELL LEUKEMIA is a lymphoproliferative disorder characterized by pancytopenia, an indolent course, and susceptibility to infections normally controlled by the cell-mediated immune system. Infection by nontuberculous mycobacteria occurs in 5% of patients with hairy cell leukemia and is frequently disseminated with a fatal outcome.[1] In this report, we describe a patient with hairy cell leukemia whose disseminated *Mycobacterium avium-intracellulare* infection had a unique site of symptomatic involvement, the popliteal artery, that resulted in a mycotic aneurysm.

Report of a Case

The patient, a 58-year-old man, was seen because for two months he had had fever, weight loss, and progressive swelling and pain in the right calf and popliteal fossa. Nine years before admission, he had suffered a right leg injury requiring several reconstructive procedures. Four years before admission, mild pancytopenia was noted, and hairy cell leukemia was diagnosed by bone marrow examination that showed typical hairy cells containing tartrate-resistant acid phosphatase. At the time of hospital admission, he had hepatosplenomegaly and right calf tenderness with edema and erythema but no palpable mass or cord. The leukocyte count was 0.6×10^9 per liter with 0.48 neutrophils and no monocytes. The hematocrit was 0.2 and the platelet count was 104×10^9 per liter. A chest radiograph showed no pulmonary infiltrates, cavities, or effusions. The patient was anergic to intradermal skin tests. Fever occurred 12 hours after admission, and broad-spectrum antibiotics were given. Daily fevers to 38°C to 39°C (100.4°F to 102.2°F) were noted. After six days, cultures of specimens of blood, sputum, urine, and cerebrospinal fluid were all negative, and antibiotic therapy was discontinued. A pulsatile right popliteal mass developed. Cultures of specimens of blood from peripheral and right femoral veins were negative.

On the eighth hospital day a splenectomy was done. The liver, spleen, and bone marrow were infiltrated by hairy cells, but neither acid-fast bacilli nor granulomata were seen. The fever continued and, postoperatively, bacteremia with *Staphylococcus aureus* was recorded. The popliteal mass enlarged, and on the 14th hospital day, a popliteal aneurysm was resected and a saphenous vein bypass graft done. After the operation, *S aureus* was cultured from the aneurysm. On the 36th hospital day, *M avium-intracellulare* grew in cultures of the liver, spleen, and bone marrow. On reexamination of the mycotic aneurysm, acid-fast bacilli were visible microscopically (Figure 1) and *M avium-intracellulare* grew from specimens of both the aneurysm and peripheral blood.

Therapy with isoniazid, rifampin, ethambutol hydrochloride, and streptomycin was begun, but arterial hemorrhage occurred in the right popliteal space, requiring an above-the-knee amputation. The patient's daily fever gradually resolved, and on the 46th hospital day, rifampin was replaced by ansamycin, 150 mg given orally per day. Subsequent antimicrobial susceptibility testing showed sensitivity to ansamycin, 2 μg per ml; isoniazid, 5 μg per ml; streptomycin, 10 μg per ml; and cycloserine, 30 μg per ml. By the 56th hospital day, he was afebrile, and, after rehabilitation, was discharged from the hospital.

Seven months after discharge, a pulsatile right groin mass was noted, and a 3-cm pseudoaneurysm was resected from the site of the previous proximal saphenous vein graft anastamosis. *M avium-intracellulare* grew from a specimen of this material. The organism was susceptible only to ansamycin, 2

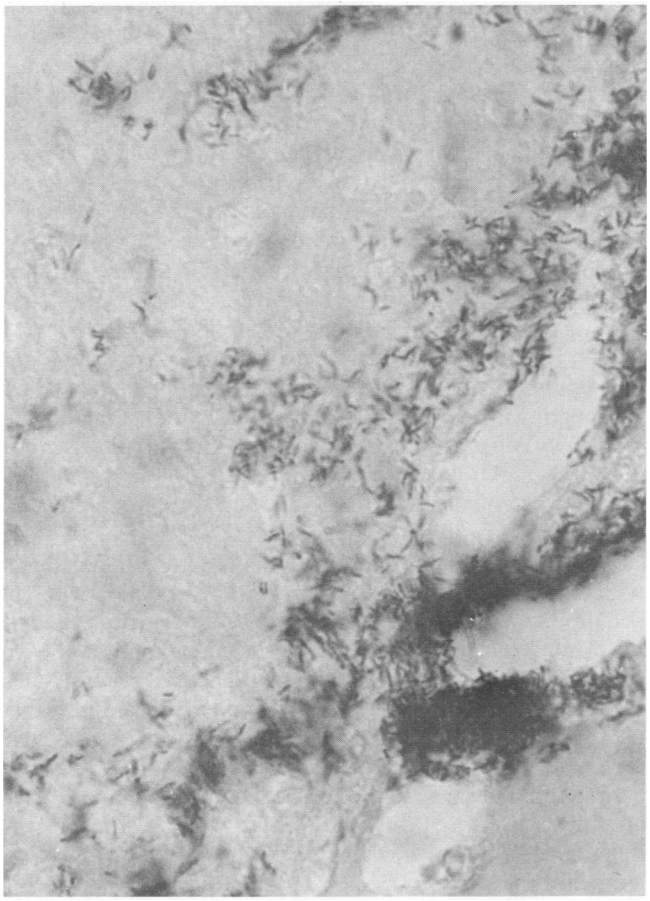

Figure 1.—A specimen of the wall of mycotic aneurysm shows acid-fast bacilli (Ziehl-Neelsen stain, original magnification × 1,000).

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μg per ml, and the streptomycin therapy was discontinued. The patient remains afebrile, and after two years of antimycobacterial therapy, the leukocyte count is 5.8×10^9 per liter, the hematocrit is 0.48, and the platelet count is 330×10^9 per liter.

Discussion

This patient is unique among other patients with hairy cell leukemia and disseminated *M avium-intracellulare* infection because he had a mycotic aneurysm caused by the organism. In the past, mycotic aneurysms were mostly a complication of endocarditis or atherosclerosis; at present they are usually a complication of trauma. In a recent series *S aureus* and *Enterobacter* were the most common pathogens,[2] though tuberculous aneurysms of the aorta have also been reported.[3,4] Approximately 1% of popliteal aneurysms are mycotic aneurysms.[5] *M avium-intracellulare* has not been previously reported as a causative agent of these aneurysms.

The source of the mycobacterial bacilli in our patient is unknown. In a recent series,[1] eight of nine patients with hairy cell leukemia and disseminated nontuberculous mycobacteriosis had abnormalities on chest radiography presumed to represent the source of infection. In contrast, our patient's lack of a demonstrable pulmonary focus together with the preexisting contaminated compound fracture of the femur raise the possibility, however speculative, that the leg was the source of infection of this saprophytic organism.

Disseminated nontuberculous mycobacterial infection occurs in 5% of all infections due to nontuberculous mycobacteria.[6] Most adults with disseminated *M avium-intracellulare* have a previously diagnosed immunosuppressive disorder,[7] and hairy cell leukemia is present in 5%. Disseminated *M avium-intracellulare* has been reported in a significant number of patients with the acquired immunodeficiency syndrome.[8] Studies of patients with malignant disease[9] and hairy cell leukemia[1,10-13] have concluded that *Mycobacterium kansasii* is the most common nontuberculous mycobacterial pathogen, though the frequency has considerable geographic variation, presumably related to the geographic distribution of the natural bacterial reservoirs. Theories concerning the predilection of nontuberculous mycobacterial infection for patients with hairy cell leukemia have focused on the nature of the host defense to intracellular pathogens. Although skin test responses are generally intact in patients with hairy cell leukemia,[13] profound monocytopenia is commonly seen. The pathologic features are often remarkable, as in the present case, for a lack of granuloma formation and a predominately granulocytic response.

Recurrence or relapse of infection is common in patients with disseminated *M avium-intracellulare* infection with the acquired immunodeficiency syndrome.[8] Local recurrence or persistence of mycobacteria at a site is uncommon, however. In our patient, persistence could be related to anatomic factors related to the presence of a thrombosed venigraft that had traversed the primary infected site, host-defense deficiency due to hairy cell leukemia, or the development of antimicrobial resistance. Because a second disseminated mycobacterial infection did not develop while the patient was being treated, and in view of his subsequent course following surgical procedures, we speculate that the anatomic factor was the most important.

Disseminated nontuberculous mycobacterial infection will develop in 5% of patients with hairy cell leukemia, and 20% of these cases are due to *M avium-intracellulare.*[1] Although mycobacterial infection is clearly not a common complication of hairy cell leukemia, it is treatable and is often a difficult diagnostic challenge. With the continued rise in the number of immunocompromised patients, infection with *M avium-intracellulare* is likely to be recognized more frequently.

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Fatal *Pasteurella multocida* Pneumonia in an IgA-Deficient Cat Fancier

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Pasteurella multocida is a ubiquitous organism well known to veterinarians as a commensal and opportunistic pathogen; it is commonly present in the mouths of healthy dogs and cats.[1] In humans, *P multocida* infections are most commonly seen by emergency department physicians who treat animal bites and scratches, but the incidence of such infections is underestimated because of a lack of clinical suspicion and of appropriate bacteriologic investigation.[2,3] The clinical manifestations of *P multocida* infections have been succinctly described by Beyt,[4] and in a comprehensive review, Weber and co-workers also divide *P multocida* infections into three groups[5]: infection from animal bites and scratches, which may progress to cause serious local complications; respiratory tract disease including pneumonia, lung abscess, and

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